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Case Report

Solitary thoracic osteochondroma causing spinal compression: Case report

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ABSTRACT

Spinal osteochondromas are very rare, and they present with nonspecific localized pain owing to bone involvement. Diagnosis is made based on direct X-ray and computed tomography (CT) imaging of the exophytic bone lesion with pedunculated or sessile structure. Although asymptomatic patients can be observed, surgical excision is the main treatment modality. We present the case of a 34-year-old man with solitary thoracic osteochondroma. The patient presented with complaints of pain in the legs, numbness, and inability to walk. The diagnosis was confirmed with CT imaging showing calcified heterogeneous bone lesion originating from the left side of T1-2 facet joint. After total excision, histopathological examination revealed the diagnosis of osteochondroma. No new clinical or radiological findings were detected in the 10-month follow-up.

Introduction

Osteochondroma is one of the benign cartilaginous tumors of the bone, which can typically be found in areas where endochondral ossification occurs. They are exophytic lesions with pedunculated or sessile structure, which develop in the peripheral parts of the epiphyseal growth plates (1, 2). Spinal involvement is rare. It constitutes 4%-7% of the primary benign spine tumors. This tumor, also known as osteocartilaginous exostosis, has 2 forms: solitary exostosis and multiple exostosis with (OD transitive form multiple hereditary exostosis-MHE) or without genetic transmission. In these tumors, where the mean age at which this disease occurs is 28.5 [2-69] years, the female/male ratio is 3:2 in solid cases, whereas there is no gender difference in patients with MHE (3). Solid form is seen in the spine in 1.3%-4.1% patients, whereas MHE is seen in 3%-9% patients (3, 4). Osteochondroma, rarely seen in the spine, originates from the posterior elements of the vertebra rather than the spine body (4, 5). It is often caused by the tip of the spinous process or transverse protrusion or the posterior elements covering the secondary ossification regions (4-6). While solid lesions involve C1 in majority of the patients, C2 involvement is most commonly observed in patients with MHE. Solid lesions are more common in the thoracic area than in the lumbar spine, whereas the incidence in the thoracic and lumbar areas of the spine is equal in patients with MHE (4, 6). They often present with regional pain according to the localization and neurological findings, such as radiculopathy and myelopathy in lesions growing into the spinal canal (1, 2). Direct radiography and computed tomography (CT) are used for tumor detection and diagnosis, whole-body bone scintigraphy is used for the detection of multiple involvement, and magnetic resonance imaging (MRI) is used for the detection of malignant transformation of the tumor and its relationship with the neurological structures (2, 3, 5). Malignant transformation rate is significantly higher in lesions associated with MHE (7). Asymptomatic cases can be observed, and patients with progressive pain complaints, compression findings, neurological symptoms, and malignant potential should undergo surgical treatment, including complete excision of the cartilaginous cap (3, 8). In patients where instability may develop after laminectomy and facetectomy, stabilization should be added to surgery (3, 4, 6). In this case report, the case of a 34-year-old male patient who was operated and diagnosed with osteochondroma is discussed.

Case Presentation

A 34-year-old male patient was admitted to our clinic with the complaints of pain in the legs, numbness, and inability to walk for the past 1 year, which had aggravated during the last 6 months, and he was diagnosed with lumbar discopathy in the external center and administered various medical/physical treatments. During the hospitalization, informed consent was obtained from the patient for the operation to be performed and the medical treatment to be given. He did not respond to any of them, and because of his increasing complaints, he underwent a contrast-enhanced MRI of the spine, which revealed a mass lesion in the T1 vertebra. On admission, he was diagnosed with American Spinal Injury Association classification D with left patella reflex hyperactivity, left Babinski positivity, and impaired tandem gait. Thoracic MRI showed calcified heterogeneous appearance (primary bone tumor) at T1-T2 level in the left facet joint extending from T1 inferior articular process and T2 superior articular process into the

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spinal canal, markedly narrowing the spinal canal and significantly pressing the cord toward the right anterolaterally and accompanied by increased signal suggestive of compressive myelomalacia in T2 (Figure 1, 2). The cap thickness was measured as 17.7 mm in preoperative axial CT (Figure 3). The preoperative electroneurophysiological examination (EMG) of the patient was reported as normal (Table 1).

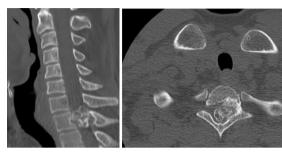


Figure 1. Sagittal and axial computed tomography imaging showing osteochondro-

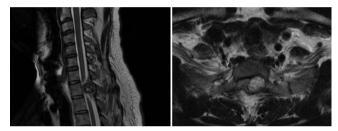


Figure 2. Sagittal and axial magnetic resonance imaging showing osteochondroma located at T1 vertebra

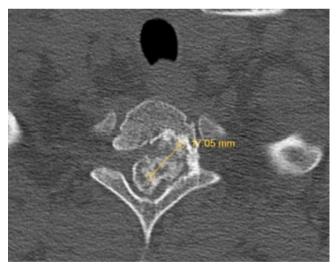


Figure 3. Preoperative axial computed tomography, osteochondroma cartilage cap [17.7 mm]

HIGHLIGHTS

- Osteochondroma is one of the benign cartilaginous tumors of the bone, which
 can typically be found in areas where endochondral ossification occurs.
- Asymptomatic cases can be observed, and patients with progressive pain complaints, compression findings, neurological symptoms, and malignant potential should undergo surgical treatment, including complete excision of the cartilaginous can
- Although it is generally considered to be benign, the combination of radical
 excision of the tumor, including cartilaginous cap owing to the local recurrence rates following insufficient surgery and the possible risk of malignant
 transformation, and the treatment practices reducing the postoperative recurrence rates is the most appropriate treatment method.

The patient was operated after the diagnosis of T1 vertebral mass and accompanied by neuromonitorization (Graph 1). The tumor invading the T1 lamina by posterior approach was completely excised, including the left T1 pedicle and the cartilaginous cap. Histopathological examination revealed T1 vertebra tumor consisting of a thin fibrous layer on the surface, basophilic hyaline cartilage in the middle, and bone trabeculae in the deep and was diagnosed as osteochondroma (Figure 4). Postoperative control thoracic CT and MRI showed gross total excision of the tumor (Figure 5, 6). The patient was discharged with a thoracic corset on the 7th postoperative day without any addi-

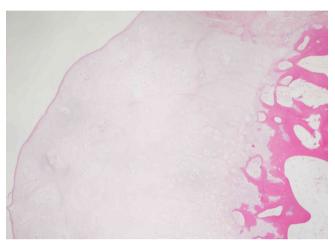
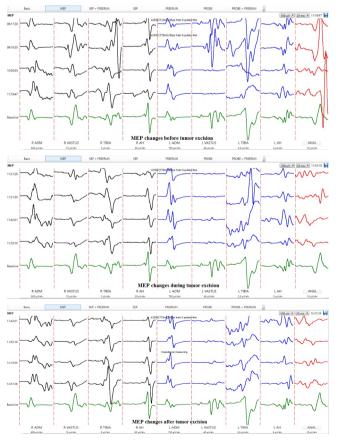


Figure 4. Hematoxylin and eosin x40 examination, a thin fibrous layer on the surface, T1 vertebra tumor consisting of basophilic hyaline cartilage and deep bone trabeculae, osteochondroma



Graph 1. MEP changes during the surgery. It is seen that there is no extra loss from MEP changes taken during surgery according to baseline in the MEP end of the surgery

Table 1. Preop ENMG						
Right/ 1. Dorsal Int.	-	-	-	-	Normal	+1 Interferans
Right/ Biceps	-	-	-	-	Normal	+1 Interferans
Right/ Tibialis anterior	-	-	-	-	Normal	+1 Interferans
Right/ Tibialis posterior	-	-	-	-	Normal	+1 Interferans
NERVE TO THINNER	DIS. LAT.	DISTANCE	AMPLITUDE	NCV		
R/D/Median (2.prm- wrist)	2,4msn	14cm	$33\mu V$	$58 \mathrm{m/sn}$		
R/D/Ulnar (5.prm- wrist)	2,3msn	13cm	$29\mu V$	$56\mathrm{m/sn}$		
R/M/Median (Abd. pol. bre.)	3,6msn	24cm	$7,8-7,7 \mathrm{mV}$	$62 \mathrm{m/sn}$		
R/M/Ulnar (Abd. dig.min.)	3,0msn	24cm	7,5-7,4mV	$65 \mathrm{m/sn}$		
R/ Peroneal superficial	2,1msn	12cm	$22\mu V$	$57 \mathrm{m/sn}$		
R/M/Peroneal (ext. dig.)	3,9msn	35cm	$5,5-5,0 { m mV}$	$53 \mathrm{m/sn}$		
L/M/Peroneal (ext. dig.)	4,4msn	35cm	3,5-3,0mV	51m/sn		

Transmission studies on the upper right and both lower extremities and needle EMG on three extremities are within normal limits.

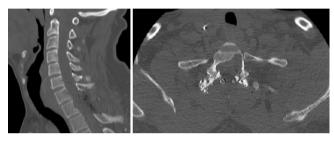


Figure 5. Postoperative first year computed tomography

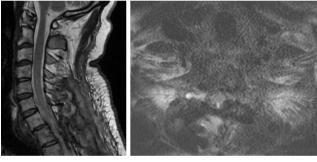


Figure 6. Postoperative first year magnetic resonance imaging

tional deficit. No new clinical or radiological findings were detected in the 10-month follow-up.

Discussion

While the solid form of osteochondromas, which are rare in spinal involvement, is as low as 1.3%-4.1%, recent studies show that solitary lesions are much more common than before (6). Solitary lesions occur more frequently in males and at a mean age of 32.8 years. Spinal osteomas, often located in the upper cervical region, originate from the tip of the spinous profess, transverse protrusion, or the posterior elements. More frequent cervical lesions have been associated with exposure of the spine to microtraumas owing to the spinal strain and mobility and with enlarged microcartilage residues in this zone (1, 2). In the patient discussed in our case report, the mass was located in the posterior elements of the T1 vertebrae in accordance with the literature (1, 2).

Patients often present with regional pain. Since the first symptom is usually localized pain, which is very common in daily practice, this may cause misinterpretations and delay in diagnosis. As the size of the tumor grows, it may present with localized pain and swelling on the affected side and rarely with compression of the spinal cord or nerve roots and neurological deficits (4, 9). Similarly, our patient was treated intermittently for neck pain. After nerve compression was

found, he was diagnosed with lumbar disc disease and received medical/physical treatment in various centers for 1 year. When he further experienced weakness in bilateral lower extremities and developed sensory defect, which involved staging, he was diagnosed with thoracic mass after additional investigations.

Although radiological evaluations alone are not sufficient for diagnosis, they can give information about spinal deformity. CT and MRI are important in evaluating the relationship between the medullary structure of the lesion and the adjacent bone segment. While CT is effective in detecting the osseous and cartilaginous margins of the lesion, MRI is particularly useful in assessing the thickness of the cartilage portion (cap). When the cartilage has a thickness of more than 3 cm, it suggests malignant transformation (10). In the preoperative MRI of our patient, the cap thickness was less than 3 cm, and no malignant transformation was considered. It should be kept in mind that there could be multiple osteochondromas, although this presentation is rare. For this purpose, the full length of the spine should be investigated preoperatively by bone scintigraphy or CT (3). In our case, preoperative scintigraphy and complete spinal CT were performed, and no second focus was detected.

One of the distinctive features of the osteochondroma is the cap part located at the end of the stem attached to the adjacent bone without cortex. Histopathologically, as in our patient, it consists of hyaline cartilage surrounded by dense collagen forming the perichondrium (10, 11) (Figure 4).

Treatment of osteochondromas is more complex because of the limited surgical access in the spine, unlike tumors in other regions, differences in vascular structures, and proximity to the spinal cord and nerve roots. Spontaneous regression was defined at a very young age in the sessile form after trauma in the pedicle form. Therefore, these benign masses can be observed in asymptomatic patients (2). Lesions which cause symptomatic or neurological deficits are treated surgically. Although the probability of recurrence is approximately 2% (2.6) in patients where the cartilaginous cap cannot be completely removed, total resection with cartilaginous cap should be targeted if possible, and spinal instrumentation and arthrodesis should be added to the treatment in patients thought to cause instability in surgical resection (12).

Malignant transformation of osteochondromas is rare. In a study conducted by Gille O et al. with a series of 150 patients diagnosed with spinal osteochondroma, malignant transformation rate was reported to be 2.5% (13). Patients with a high risk of malignant transformation, such as the thickness of the cap portion of more than 3 cm and the presence of MHE, should definitely undergo close clinical and radiological follow-up after the operation and should be evaluated with postoperative radiotherapy (RT). In our patient, because of the presence of the solitary

lesion, the thickness of the portion of tumor cap on preoperative CT being less than 3 cm, the total excision of the mass, and the absence of local recurrence or a new mass with different localization as detected in the postoperative follow-ups, the possibility of malignant transformation was low; thus, a postoperative RT was not planned.

Conclusion

Patients with osteochondroma presenting with nonspecific neck, back, and low back pain, especially in young males, often in the long bones, and, more rarely, with spinal involvement, should definitely be considered in the differential diagnosis. Although it is generally considered to be benign, the combination of radical excision of the tumor, including cartilaginous cap owing to the local recurrence rates following insufficient surgery and the possible risk of malignant transformation, and the treatment practices reducing the postoperative recurrence rates (clinical/radiological follow-up and RT) is the most appropriate treatment method.

Informed Consent: During the hospitalization, informed consent was obtained from the patient for the operation to be performed and the medical treatment to be given.

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Conflict of Interest: The authors have no conflicts of interest to declare

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References

- Quirini GE, Meyer JR, Herman M, Russell EJ. Osteochondroma of the thoracic spine: An unusual cause of spinal cord compression. Am Soc Neuroradiol 1996: 17: 961-4.
- Thiart M. Lumbar osteocondroma causing spinal compression case report and review of the literature. SA Orthop J 2010; 44-7.
- Ofluoğlu AE, Abdallah A, Gökcedağ A. Solitary osteochondroma arising from cervical spina bifida occulta. Case Rep Orthop 2013; 2013: 509745. doi: 10.1155/2013/509745. Epub 2013 Dec 8. [Crossref]
- Çaylı SR, Irkkan Ç, Sökmen Ö. An unusual presentation of solitary osteochondroma of the cervical spine: Case Report. Turk Neurosurg 2001; 11: 65-8.
- Nitin Samal G, Chavan M, Badole CM, Pisulkar G, Shashikant. Osteochondroma arising from spinous process of lumbar spine without spinal cord compression: A Rare Presentation. Int J Multidisciplinary Health Sci 2014; 1: 23-8.
- Bess RS, Robbin MR, Bohlman HH, Thompson GH. Spinal exostoses: Analysis of twelve causes and review of the literature. Spine 2005; 30: 774-80.
 [Crossref]
- Günay C, Atalar H, Yıldız Y, Sağlık Y. Spinal osteochondroma: A report on six patients and a review of the literature. Arch Orthop Trauma Surg 2010; 130: 1459-65. [Crossref]
- Huda N, Julfiqar M, Pant A, Jameel T. Giant cervical spine osteochondroma in an adolescent female. J Clin Diagn Res 2014; 8: LD01-2. [Crossref]
- Srikantha U, Bhagavaluta ID, Satyanarayana S, Somanna S, Chandramouli BA.
 Spinal osteochondroma: Spectrum of a rare disease. J Neurosurg Spine 2008; 8: 561-6. [Crossref]
- Thakur NA, Daniels AH, Schiller J, et al. Benign tumors of the spine. J Am Acad Orthop Surg 2012; 20: 715-24. [Crossref]
- Sansur CA, Pouratian N, Dumont AS, Schiff D, Shaffrey CI, Shaffrey ME. Part II: Spinal-cord neoplasms-primary tumours of the bony spine and adjacent soft tissues. Lancet Oncol 2007; 8: 137-47. [Crossref]
- Öztürk C, Tezer M, Hamzaoğlu A. Solitary osteochondroma of the cervical spine causing spinal cord compression. Acta Orthop Belg 2007; 73: 133-6.
- Gille O, Pointillart V, Vital J-M. Course of spinal solitary osteochondromas. Spine (Phili Pa 1976) 2005; 30: E13-9. [Crossref]